National SCIENCE Challenges

A BETTER START

E Tipu e Rea

A Better Start

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Brief Evidence Reviews for the Well Child Tamariki Ora Programme

Report submitted to MoH on 11 December 2019

Whakapūpūtia mai ō mānuka,
kia kore ai e whati

Cluster the branches of the manuka,
so they will not break

Foreword

The Ministry of Health is responsible for the development of policy advice on children's health and the future direction of the Well Child Tamariki Ora (WCTO) programme. The WCTO programme is the universal health service in New Zealand, which is responsible for protecting and improving the health and wellbeing of children from birth to 5 years of age. This is achieved through health and development screening and surveillance, whānau care and support, and health education.

The current programme is based on the evidence available at the time of the last programme update in 2007. Therefore, the Ministry of Health is reviewing the current WCTO Framework and associated Schedule (developed in 2002) to ensure that WCTO services meet the current needs of children and their whānau, and address the issues they face. The present review was initiated in 2019 and is the second review of the programme, as the first was carried out in 2006. In preparation for this review, the Ministry of Health has commissioned an evaluation of the recent literature on some of the new and emerging issues for preschool children, as well as possible ways to address them.

The purpose of this review includes ensuring that the programme is underpinned by the latest research and evidence. This is particularly pertinent to the current Schedule of Universal Contacts delivered, and one of the work-streams of the review is to consider the timing, content, and intensity of the Schedule, and associated additional contacts. This work stream will support the development of an integrated framework of universal wellbeing contacts for the pregnancy to 24 years of age life course.

The Ministry of Health require the brief evidence reviews (BERs) to synthesise relevant evidence about what works in key areas for children, including development, vision, hearing, emotional and mental health, and growth. The BERs adopted the He Awa Whiria – Braided Rivers approach and include consideration of what will work for Māori tamariki and whānau, and Pacific children and families within each domain. The BERs have helped to identify any knowledge gaps where further work and research may be needed, to inform further development of the WCTO programme.

The WCTO review is a key health contribution to the Government's Child and Youth Well-being Strategy. It forms part of the Ministry of Health's work programme to transform its approach to supporting maternal, child, and youth well-being.

The Ministry of Health have commissioned A Better Start: E Tipu E Rea National Science Challenge to undertake 11 health related BERs that will inform the WCTO review and decision making on the future core service schedule, and additional health and social services for children in New Zealand. The aim of the BERs is to ensure that decisions are grounded in, and informed by, up-to-date evidence. BERs are intended to synthesise available evidence and meet time constraints of health care decision makers. Internationally health technology agencies have embraced rapid reviews, with most agencies internationally offering these alongside standard reviews. These 11 BERs that we have conducted have been performed in a very short time which was a very challenging task.

A Better Start is a national research programme funded by the Ministry of Business Innovation and Employment (MBIE). The objective of A Better Start is to improve the potential for all young New Zealanders to lead a healthy and successful life. To achieve this, A Better Start is researching methods and tools to predict, prevent, and intervene so children have a healthy weight, are successful learners, and are emotionally and socially well-adjusted. A Better Start consists of more than 120 researchers across 8 institutions.

The BERs cover 11 domains critical to the WCTO programme, which are: neurodevelopment (#1); parent-child relationships (#2); social, emotional, and behavioural screening (#3); parental mental health problems during pregnancy and the postnatal period (#4); parental alcohol and drug use (#5); excessive weight gain and poor growth (#6); vision (#7); oral health (#8); adverse childhood experiences (#9); hearing (#10); and family violence (#11). The BERs have synthesised relevant evidence about what works in key areas for children across these domains, which were assessed with careful consideration of what will work for Māori tamariki and whānau and Pacific children and families. They have also identified knowledge gaps where further work and research may be needed to inform further development of the WCTO programme.

Within each domain, a series of 6–14 specific questions were drafted by the Ministry of Health, and subsequently refined with input from the large team of researchers assembled by A Better Start. A Better Start established discrete writing teams to undertake each BER. These teams largely consisted of a post-doctoral research fellow and specialty expert, often in consultation with other experts in the field. Subsequently, each BER was peer reviewed by at least two independent experts in the field, as well as two Māori and a Pacific senior researcher. In addition, senior clinical staff from the Ministry of Health have reviewed each BER. These were then revised to address all the feedback received, checked by the editors, and finalised for inclusion in this report.

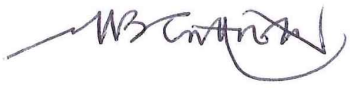
Whilst each of these domains are reviewed as discrete entities, there is considerably inter-relatedness between them. In particular, neurodevelopmental problems can be impacted by parent-child relationships, parental mental health, and pre- and postnatal drug exposure. Similarly, children who have problems with growth, vision, or oral health may also have neurodevelopmental disorders.

Most of the evidence available for these BERs comes from international studies with limited data from New Zealand, in particular there is limited information about Māori, Pacific, and disadvantaged families. These are the tamariki and whānau in whom the WCTO Programme services are more scarce, yet could potentially offer the greatest benefit.

The criteria for screening include the requirement for an effective and accessible intervention; the corollary is that screening should not be offered if there is no benefit to the individual being screened. The essential issue is therefore to identify those infants and preschool children and their whānau who would have better outcomes following intervention; this includes better outcomes for the whānau.

The current WCTO programme has had a greater emphasis on surveillance rather than screening. Many of the questions in the BERs address screening. A change in the WCTO programme that further extends into screening will require substantial upskilling of many WCTO providers, as well as redirection of resources. Importantly, Māori and Pacific iwi and community views must be considered before any new screening programmes are to be included.

It should be noted that a shift towards screening rather than surveillance may prevent health and behavioural problems. The economic benefits of prevention and early intervention are well documented, with early interventions showing that for every dollar spent there are substantial savings to health, social services, police, and special education resources.



Professor Wayne Cutfield
Director of A Better Start National Science Challenge
On behalf of the editors, authors and reviewers of the brief evidence reviews

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10 Hearing screening in childhood (excluding newborns)

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List of Figures and Tables

Disclaimer

This brief evidence review was commissioned by A Better Start National Science Challenge (the Challenge) on behalf of the New Zealand Ministry of Health. It was prepared over a relatively short time based on the evidence available to the authors at the time of its preparation. The authors have made considerable efforts to perform a comprehensive and balanced evaluation of the existing evidence. However, this brief evidence review cannot be considered an exhaustive analysis of the existing peer-reviewed and grey literature on the topic, and it may not reflect the potentially conflicting views of all experts in the field. There could have been important omissions, and additional evidence might have also come to light since completion of this final draft. Thus, this brief evidence review should be considered with the appropriate caution. A previous version of this document was peer-reviewed by Māori and Pacific researchers and by independent experts in the field. Peer reviewers were anonymous, unless they have otherwise been identified by name. Please note that this brief evidence review does not represent the views of the Challenge or the Ministry of Health; rather, it reports the independent conclusions of the listed authors.

Conflicts of interest: The authors have no financial or non-financial conflicts of interest to declare that may be relevant to this work.

Aims

This rapid review attempts to answer the following questions about childhood hearing screening as posed by the Ministry of Health.

Review Approach

A literature review was performed using Scopus and Google Scholar. The search was conducted using combinations of the following terms: hearing screening, preschool, early childhood, otoacoustic emissions, sweep test, Pacific, Māori. Key references from identified articles were also included where appropriate. The search was limited to studies published in English.

10.1 What are the most common hearing impairments in early childhood (0-5 years) in New Zealand, and what is their prevalence?

Despite the availability of universal newborn hearing screening there are a number of children who are lost to follow-up each year, further some children who arrive in the country as immigrants may not have been screened; also there are a number of causes of late / delayed onset hearing loss, including middle ear disease, but also slight or progressive sensorineural hearing losses which are not detected through the newborn programme and acquired hearing loss

New Zealand specific prevalence values are not available, although estimates can be made from data obtained through the NZ Deafness Notification Database (NZDNDB), B4 school check data and census data. These data are incomplete however, with NZDNDB data estimated to reflect only 50-70% of permanent hearing loss diagnosis every year[1], the B4 school check data has incomplete coverage and is conflated with referrals due to otitis media, and census data are dated and based on parental interpretation of "disabling hearing loss".

NZDNDB data indicates that 88% of reported cases have an unknown cause.
From 2010-2017, 70% of notifications were for bilateral hearing loss, the remaining 30% were for unilateral losses with severe unilateral losses called "Single Sided Deafness" (SSD) accounting for 6% of notifications. 40% of cases were coded as likely present since birth, 14% of cases unlikely to have been present since birth and 46% of cases of unknown duration. There are no data or information regarding the proportion of cases of hearing loss that are progressive in nature. The severity profile of hearing loss reported in the NZDNDB is summarised in Figure 10.1.

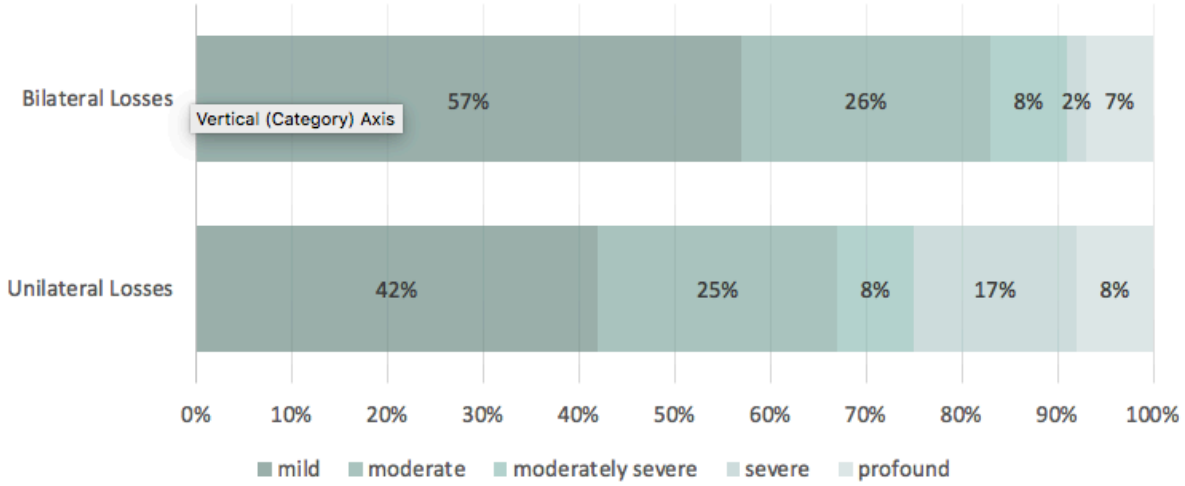

Figure 10.1. Unilateral and bilateral hearing losses by degree reproduced from Deafness Notification Report (2017) Hearing loss (not remediable by grommets) in New Zealanders under the age of 19; Figure 13, page 49[1].

The B4 School Check is a nationwide programme which offers free hearing screening for all 4 year olds and aims to detect mild losses or poorer. Coverage has improved significantly in the last 10 years although it varies significantly by ethnicity (see section 10). The following table has been replicated from the NZDNDB report as it provides insight to the current B4 school screening programme. Referrals do not necessarily indicate a permanent hearing loss and may include referral due to transient middle ear disease or false positive results.

Table 10.1. B4 School Check Hearing Screening Data reproduced from Deafness Notification Report (2017) Hearing loss (not remediable by grommets) in New Zealanders under the age of 19; Table 15, page 40[1].

Outcome	Description	2010/11	2012/13	2014/15	2016/17
Pass Bilaterally	The child was screened and passed	58%	71%	79%	81.2%
Referred	The child was screened and referred to a relevant service	5%	5%	5%	5.2%
Rescreen	The child was unable to complete the screen, so a rescreen was booked, normally in around 6 months.	7%	7%	6%	4.8%
Under care	The child is already under the care of a relevant service	1%	3%	3%	3.5%
Decline	The hearing check was declined by the caregiver	4%	4%	1%	0.7%
Not Checked	The child did not receive a hearing check	24%	11%	6%	4.5%
Population	Derived from PHO enrolled populations	63,585	64,911	63,730	62,581

Census-derived data are old (2001/2), and were collected prior to the advent of the NZ Universal Newborn Hearing Screening and Early Intervention Programme (UNHSEIP). They show prevalence of hearing loss of 1.7%, 2.7% and 2.0% for children aged 0-4y, 5-9y and 10-14y respectively[2].

Given the lack of NZ specific information, prevalence data from similar countries may provide better information.

British evidence suggests that there is a significant increase in prevalence of hearing loss (>20 dB HL) from birth (2-3 per 1000) to school age (6-10 per 1000), with prevalence continuing to increase between ages 6 to 8[3]. This trend remains true with more recent data looking at mild or greater hearing losses with prevalence increasing from 1.79 per 1000 at birth to 3.65 per 1000 for children at school entry[4]; and again if only looking only at hearing losses greater than 40 dB HL (1.06 per 1000 at birth rising to between 1.65-2.05 per 1000 among children age 9 years or older)[5].

This is consistent with NZ UNHSEIP data which reports 1.2 cases of bilateral hearing loss per thousand babies and an additional 1.1 cases of unilateral hearing loss per thousand babies screened. This places the NZ prevalence at birth slightly higher than that of the UK data. Sixty percent of diagnoses in the 2017 NZDNDB are attributable to New Zealand's UNHSEIP. We therefore might infer that 40% of reported losses are either late notifications, UNHSEIP misses, progressive or acquired losses. Note however that NZDNDB reporting for older children is likely less reliable and significantly underrepresents the proportion of children as evidenced by international data.

Note from the UK data this increase in prevalence with age is primarily due to sensorineural hearing loss, and likely a combination of losses that were too mild to be picked up at birth, progressive losses, and adventitious hearing loss (e.g. CMV, measles, mumps, meningitis). UK findings indicate that for every child detected through newborn hearing screening programmes another 50-90% more children will be detected with permanent sensorineural hearing loss by age 9[5].

Another significant contributor to the increase in hearing loss prevalence during childhood is Otitis Media with Effusion (OME), which can cause transient, chronic and permanent hearing losses. From international data approximately 90% of children have OME at some time before starting school[6], and 25% of school aged children may have effusion at some time during the year[7]. While OME is highly

prevalent it will spontaneously resolve in most children within 3 months[8,9]. Therefore, a period of watchful waiting is recommended before any medical intervention is applied. However, 30 – 40% of children will have recurrent OME, and 5-10% of episodes last one year or longer;[6,8-10] and if middle ear effusion is present for longer than three months there is little chance of recovery without medical treatment[6,8-10]. The degree of hearing loss associated with OME varies from minimal to moderate (15 – 50 dBHL across 0.5 – 4 kHz); therefore care needs to be taken while screening to detect persistent OME but to avoid over-referring for transient cases, which can create unnecessary burden upon families and health care services.

10.2 What are the long-term consequences of undiagnosed hearing impairments?

There is evidence that children with unrecognized and unmanaged unilateral or minimal bilateral hearing loss have significant speech-language delays, negative educational consequences, and behavioural problems[11-13]. The greater the degree of loss the more significant the long-term impact on the child and their future vocational attainment[14]. In the case of chronic middle ear disease, long term sequelae include progressive hearing loss, eardrum perforation, sensorineural deafness, balance disorders, mastoiditis, and meningitis.

10.3 Behavioural or objective screening – what is the most appropriate tool to detect hearing impairments in children aged 0-5 years beyond birth?

10.3.1 Overview

Outlined below are methods that have been investigated as screening tools for hearing loss in the preschool population. We have excluded tools that could be used for middle ear disease but are not sensitive or specific for hearing loss (e.g. Immittance Testing). Generally, much of the current literature revolves around the use of pure tone audiometric screening (behavioural testing) and the use of otoacoustic emissions (OAE; objective testing based on physiological activity in the normally-functioning inner ear). Three systematic reviews[3,15,16] have covered this topic and concluded that with the available (limited) evidence pure-tone screening had higher sensitivity than OAE testing in school age children, however for preschool children aged 4 the difference in sensitivity between the two tests has not been adequately investigated and is a matter of debate;[17,18] and for 3 year-olds pure-tone screening is not recommended as most children are unable to perform the test reliably at this age[16]. This has led to the recommendation of the use of OAEs in children chronologically and developmentally under 3 years of age by the American Academy of Audiology (AAA)[16]. More recently a series of papers have encouraged the use of Distortion Product Otoacoustic Emissions (DPOAEs; a subgroup of OAEs that provide a degree of specificity about the frequencies at which hearing losses may be present) to be re-examined as a screening method[17]. This work addresses many of the identified limitations of OAE screening (i.e. reduced sensitivity for mild losses, insensitivity to auditory neuropathy dysynchrony disorder, and difficulty to obtain low frequency results) and proposes a screening protocol that may be more efficient than behavioural screening because of its speed, frequency specificity and the need for less cooperation from the child.

Emergent mobile app based technologies (e.g. Hear Screen[19-21], SoundScouts[22] and Digits in Noise Tests[23,24]) are also discussed, all of which fall within the behavioural testing approach to screening.

Questionnaire based screening approaches were also investigated[25,26], however a recent rapid review found insufficient evidence that parent- or teacher-completed questionnaires can reliably be used to screen for hearing loss[27].

According to the AAA(2011) guidelines[16], an effective screening tool should correctly identify 90-95% of children who have existing hearing loss (sensitivity), and should fail no more than 5-10% of children with normal hearing (specificity). There is a wide range of sensitivity and specificity values for both behavioural and objective screening approaches (see below).

10.4 Behavioural testing (manual and automated pure-tone screening, digits in noise tests)

10.4.1 Pure tone audiometry screening

Screening using pure-tone audiometry or the Pure-tone Sweep Test is the current method used in New Zealand[28]. It has traditionally been considered the gold standard in screening for school aged children[16]. The current methodology uses a manual approach, although automated and app based methods are now available. App based screening is still in development and not commonly used internationally for preschool children. The advantage of a manual approach is that is allows flexibility when working with this age group[15].

Administration

Manual Pure-Tone Audiometric Screening

Using calibrated headphones with a screening audiometer, children are required to respond to a tone by performing a task (e.g. placing a peg on a pegboard). Responses are checked for a set of frequencies (e.g. 0.5, 1, 2, and 4kHz) at a specified sound level (e.g. 20 dBHL).

Screening times are usually at least 4-5 minutes. However in best case scenarios test times are 45 seconds for instructions and then a further 60 seconds for the actual screen[29].

Automated PTA Screening

This can be performed using specialised screening audiometers or using an App on mobile phones or tablets with calibrated headphones[30,31]. App based methods also monitor background noise levels to enhance test reliability. Screening times tend to be faster (around 12.3%) than manual approaches, and reliability is comparable for older children (7-9 year olds)[19].

Accuracy

Sensitivity and specificity compared to pure-tone audiometry performed in a sound treated room varies significantly across studies from 50% - 93% sensitivity and 70%- 99% specificity (Table 1). A number of papers present data indicating that automated app based testing is comparable in sensitivity and specificity to manual testing[19-21].

Table 10.2. Reported Sensitivity and Specificity of Behavioural Pure-tone Screening Studies performed in a real world setting.

Source (n) [age]	Test evaluated	Definition of screening fail	Reference standard	Sensitivity	Specificity
Sabo et al., 2000[32] (583) [5-9y]	Pure tone sweep test	>25 at 0.5 kHz and >20 dB at 1,2, and 4 kHz	PTA	87%	80%
Holtby et al., 1997[33] (610) [5-6y]	Puretone sweep test	No response at 20 dB in either ear at any frequency	PTA and Tympanometry	86%	70.2%
Fortnum et al., 2016 [34] (240) [4-6]	Puretone sweep test	No response at 20 dB in either ear at any frequency	PTA	89%	78%
Fortnum et al., 2016 [34] (240) [4-6]	Automated Handheld screener	>20 dB HL at 1 kHz and >35 dB HL at 3 kHz	PTA	83%	83%
FitzZaland and Zink, 1984[35] (3510) [4.5- 7y]	Puretone sweep test	>25 dB at 0.5 and 4 kHz , and >20 dB at 1 and 2 kHz	PTA and tympanometry	93%	99%
Halloran et al., 2009[18] (1061) [3-19y]	Puretone sweep test	>20 dB at 1, 2 or 4 kHz	PTA	50%	78%
Kam et al., 2014[30] (6231) [3-7y]	Automated test using tablet and noise cancelling phones	>30 dB	PTA (959)	3y: 33% 4y: 54% 5y: 92% 6y: 95%	15% 32% 79% 100%
Mahomed-Asmail et al., 2016[19] (1070) [8y±1.1y]	Smartphone hearing screening using the hearScreen™	>25 dBHL at 1, 2, and 4 kHz	PTA	75%	98.5%
Dillon et al., 2018 (116) [4y-14y]	Game based screening using the SmartScreen App	>20 dBHL at 0.5, 1, 2 or 4 kHz	PTA	86%	93%

Note that age had a significant impact on sensitivity and specificity values and where published, age is shown in the first column. All screening tests were conducted in real-world (non-sound-treated) conditions. Sensitivity and Specificity are relative to pure-tone audiometry conducted in a sound-attenuating chamber, but diagnostic criteria for hearing loss were not reported.

Limitations

Traditional PTA screening requires a high level of expertise and training[16], although this is not the case for newer automated procedures[36]. Screening of children younger than 3 years is unreliable with behavioural techniques, as is screening of developmentally delayed children[37] who may not understand or be able to perform the task.

Background noise is a significant issue and guidelines recommend levels no louder than 40-45dBA. Keeping background levels down to this level significantly reduces false positive rates by as much as 60%[35]. However, levels this low in a real world context are difficult to achieve; automated techniques and approaches that incorporate real time background noise monitoring and noise cancellation may help mitigate this limitation.

10.4.2 Digits in noise test (speech in noise screening)

The digits in noise test developed by Smits et al. (2004)[23] is a behavioural screening tool that relies of the loss of sensitivity to speech stimuli in noise with hearing loss. It is a closed set automated adaptive

speech in noise screening test using combinations of 3 digits (triplets) as speech material. The test measures speech reception threshold (SRT) within noise. The SRT is compared to set pass / fail criterion. Sensitivity and specificity in adults are around 80 to 90% to distinguish between normal hearing and hearing impaired listeners when compared to pure-tone audiometry conducted in a sound booth[24,38]. The Digits in Noise Test has been used in school screening programmes however it is not currently as specific as other screening methods mentioned above for a school population and was assessed only with older children (ages 9 – 16)[24]. Younger children are able to perform the task (age 5), however we have not found any sensitivity or specificity data or validation for this age group in a screening context[39]. Advantages of this test are that it does not require calibrated headphones and can be performed on a mobile phone or over the internet because it responds to the relative sound levels of the digits and the noise in which they are presented. It may also detect hearing damage before it becomes evident as reduced hearing thresholds on an audiogram. As such the poorer specificity data may reflect greater sensitivity of this test to hearing loss that is not yet detectable on a pure-tone audiogram. Speech in Noise Tests measure the relative sound level of speech to background noise and so are less sensitive to conductive losses than tests of absolute hearing level[40]. This is useful if the purpose is not to test for transient middle-ear disease, but a limitation if the screening programme is aimed at detecting these.

10.4.3 App / Game-based Screening (Sound Scouts)

Sound Scouts is a game-based hearing test delivered over the internet or via App, it can be downloaded and used without the involvement of a clinician, for children down to age 4.5 years[22]. Of note, it is currently available online as a screening tool for school age children with support from the Australian Department of Health.

Sound Scouts incorporates 3 separate hearing tests / games; a test of speech in quiet and noise, and a test of tones in noise[22]. It has been evaluated in a single piece of published research (in 116 children). In the study 8.6% of children were unable to perform the task reliably.

Duration of testing is approximately 15 minutes, including a five minute setup period which involves a supervising adult. Testing needs to take place in a quiet room.

Sensitivity and specificity are comparable to other screening approaches at 0.5, 1, 2, and 4 kHz (**Error! Reference source not found.** and **Error! Reference source not found.**). In the study, the cases of hearing loss missed by the test were all mild hearing losses up to 30 dBHL.

10.5 Objective Testing

10.5.1 Otoacoustic Emissions (OAEs)

Otoacoustic emissions (OAEs), are low amplitude signals generated in a normally-functioning ear by the outer hair cells of the cochlea in response to a sound stimulus. The presence of OAEs indicates that the pre-neural cochlear receptor mechanism and middle ear mechanism can respond to sound in a normal way. OAEs come in two primary forms, Transient Evoked (TEOAE) or Distortion Product (DPOAE). For screening, TEOAEs are produced by the presentation of a relatively high-level (80-86 dB pSPL) click stimulus. With current protocols TEOAEs are expected to be present in ears with normal hearing sensitivity and absent in cases of mild hearing loss (>35dBHL)[41]. They are sensitive to conductive pathologies, however they are less sensitive than tympanometry. Using click stimuli TEOAEs can detect hearing losses between 1-4kHz. There is good evidence that OAE testing is a useful tool for screening within the paediatric population[15,17,42,43].

The majority of research on the efficacy of OAEs as a screening tool has used TEOAEs and this therefore dominates the systematic reviews on screening tools discussed above[3,15,16], however more recently there has been an emphasis on the utility of DPOAEs for this role[44-47]. Distortion Product otoacoustic emissions are typically recorded using a series of paired tones between 1 – 6kHz, although it is possible to record up to 10kHz[16]. . The presentation of paired tones results in the generation of a third (lower frequency) tone by the hair cells within the cochlea. This means that DPOAEs recorded using high frequency stimuli are sensitive to middle ear disease and conductive losses that primarily affect low frequencies[48]. DPOAEs are less sensitive to hearing loss than TEOAEs and can be detected in some cases with 40-60 dBHL of hearing loss depending on the protocol used[41]. This statement however does not consider the amplitude of the DPOAE which is also affected by hearing loss. Hall (2016)[17] indicates that OAE sensitivity can be improved to detect hearing losses greater than 20dBHL by looking at both the noise floor (detectability) and amplitude of the DPOAE. This is currently done in diagnostic assessments of hearing loss but has not been implemented within a screening programme.

The advantages of OAEs for paediatric populations are many: results are not affected by age, cognitive level and language. Furthermore, results may be less susceptible to background noise levels than pure-tone audiometric screening (depending on protocol, equipment, and coupling method used). Testing is generally quick (within 30 seconds using an optimised method),[17] although other studies have reported longer test times ranging from 25-330 seconds with TEOAEs;[16] and a median time of 4.8 minutes (range:1 min – 30 minutes) to complete visual inspection and DPOAE screening of both ears on preschool children[49].

Accuracy

There are many studies that examine the accuracy of OAEs as a screening tool, however none incorporate the latest recommendations from Hall (2016). Few compare OAEs to the gold standard of diagnostic pure tone audiometry (PTA), with most referencing a puretone screen with or without tympanometry.

The following table is an adaptation of the work of Bamford et al. (2006)[3], Prieve et al. 2015[15] and Strabrawa & Scott (2019)[50] it includes only studies in which screening was performed in a real world setting.

Table 10.3. Reported Sensitivity and Specificity Values for OAE studies performed in a real-world setting

Source (n) [age]	Test evaluated	Definition of screening fail	Reference standard	Definition of hearing impairment	Sensitivity	Specificity
Sabo et al., 2000[32] (583) 5-9y	TEOAE	Response of 3 frequencies ≤3 db SNR, min 70%	PTA	Not Reported	65%	91%
Nozza et al., 1997[51] (66) [5-10y]	TEOAE	Various*	PTA	Not Reported	67-100%*	80-98%*
Taylor and Brooks, 2000[52] (152) [3-8y]	TEOAEs Tympanometry Screening	Response of 3 frequencies ≤3 db SNR	Pure tone sweep test	PTA >20 dBHL at 1, 2 and 4 kHz	81%	94%
McPherson and Smyth, 1997[53] (150) [5-13y]	TEOAE	Various*	PTA	PTA >15 dBHL at 0.5, 1, 2, and 4 kHz	84% 70% 78% 57%	53% 88% 81% 93%
Driscoll, Kei and Macpherson, 2001[54] (940) [6y]	TEOAE Tympanometry	Various*	Pure tone sweep test	PTA >20 dBHL at 1,2 and 4 kHz	70-89%*	84-96%*
Yin et al., 2009[55] (744/142*) [2-6y]	TEOAE		Pure tone sweep test (142 participants)	PTA >25 dBHL at 1,2 and 4 kHz	100%	94%
Lyons et al., 2004[56] (1003) [4.1-7.9y]	DPOAE + Tympanometry	Various DPOAE* SNR criteria and normal tympanogram	Pure tone sweep test	PTA >25 dB at 0.5, 1, 2 and 4 kHz	97%* 97% 98% 96%	86%* 83% 74% 95%

** indicates that the pass/refer criteria were varied systematically in order to show how they could be set. In general, settings that increased sensitivity reduced specificity and vice versa.*

Administration

OAE testing is performed by the insertion of a small speaker and microphone (probe tip) into the ear. Screening OAE machines are handheld devices and some can perform tympanometry (testing of the mobility of the tympanic membrane and middle ear status) as well. The child will hear an audible click (TEOAEs) or tones (DPOAEs). Results for each ear are generally obtained within 30 seconds during which the child needs to stay still and quiet; restlessness will cause the test to take longer. Results are presented to the screener as a simple pass or refer.

Limitations

Limitations of OAEs as a screening tool are discussed in the AAA (2011)[16] guidelines and subsequently addressed by Hall (2016)[17]. They are as follows:

It is difficult to record OAEs in the low frequency range (<1000 Hz) due to contamination from physiological and ambient noise (the same issue applies to Screening PTA) and as discussed above

DPOAEs tested at mid frequencies are still sensitive to low frequency conductive losses. Hall (2016)[17] recommends focussing testing on a frequency region of 2-5kHz to avoid low frequency interference.

OAEs are insensitive to Auditory Neuropathy Spectrum Disorder (ANSD); as OAEs are a pre-neural response any hearing losses that originate at or after the sensory hair cell and auditory nerve synapse will not be detected. However, the chances of a *miss* due to this is remote as almost all children with ANSD should be detected at birth through the newborn hearing screening programme, which uses evoked neural response testing. Additional screening questions can be used to further mitigate the chances of a *miss* by asking if the child was admitted into NICU at birth or has a sibling with a hearing loss[17].

DPOAEs are less sensitive to hearing loss when only signal to noise ratio (SNR) is used as a pass/fail criterion. However, addition of a secondary criterion of amplitude can increase DPOAE sensitivity significantly. Hall (2016)[17] proposes a criterion of =>0dB SPL amplitude and SNR of => 6dB. This proposal has not yet been evaluated.

10.5.2 Immittance Testing

Immittance Testing in the form of tympanometry is a measure of ear drum movement and is sensitive to some forms of conductive hearing loss including Otis Media with Effusion. However; it is insensitive to sensorineural hearing losses. Tympanometry and acoustic reflex testing have an important role in determining pathology and as they are very quick to administer have a useful adjunct role in the screening process as they help to determine likely cause of a refer result and therefore appropriate referral pathways[57].

10.5.3 Auditory Evoked Potentials

AABR testing as performed in the newborn hearing screening programme is not appropriate for this age group as it requires the child to be asleep, other auditory evoked potential methods such as ASSR or cortical testing are not currently viable screening methods due to long test time duration.

10.5 Summary

Both Pure-tone Screening and OAEs are useful tools for hearing screening. Pure-tone screening is only viable for testing at age four and above. OAEs can be used at all ages. Digits in noise testing is a viable screening tool for older school children but is still in development. Based on a single study, game based screening appears to be a useful tool for screening school aged children, though an effective approach to programme delivery is needed. Auditory evoked potentials are not a viable approach for wakeful children.

10.6 What is the optimal time, or times, to conduct a hearing screening test?

There is some evidence suggesting that more frequent testing is beneficial in the preschool population, particularly for high risk, and poorer populations[58].

Screening of children throughout primary and intermediate school has been advised in a report by the American Academy of Audiology (AAA), based on screening data from over 200,000 children from three schools in the United States[12]. They found that 3-6% of children screened were referred. Their results indicated that a single screen at 4 years of age would identify only 25-50 % of the newly detectable hearing losses. (AAA, 2001, pg. 18)[16]. The AAA therefore recommend screening at ages 3-4 (preschool), 5, 6, 7, 10 and 12 or 14 at a minimum[16]. These guidelines may place too much focus on detection of hearing loss by screening, and there are other concerns regarding the cost of implementing such an extensive programme. However, there is no data available to address these issues.

Other organisational guidelines recommend screening after the neonatal period because of the significant increase in prevalence of hearing loss up to age 9,[4] including the latest Joint Committee on Infant Hearing Position Statement[59].

A recent study comparing two districts with and without school screening found no benefit in cost effectiveness for school entry screening (SES)[34]. With children living in the district without SES being detected slightly earlier and detection rates being comparable to the district with SES. Note however the district without SES made use of a well-established ad-hoc referral system, in which referral was driven by parental, preschool teacher, and GP concern.

Further, the district with an SES programme had a lower referral rate to hearing services. This is an important finding and directly relates to the economic effectiveness of such programmes. The authors of the study concluded that there are two ways in which SES may be cost-effective, either a reduction in the number of referrals associated with SES or an increase in referrals due to a lack of SES. Note for example that the referral rate for the SES programme studied was 10.6%, which is over twice that of the NZ B4 School check[1]. The authors commented that caution should be taken in interpreting their results as they are not necessarily generalisable, and if withdrawal of school based screening is to be considered it needs to be carefully managed to ensure that an ad-hoc referral system is working effectively[3.] This is particularly important in NZ as evidenced by NZDNDB data in which parents were 3rd most likely to suspect hearing loss behind Vision Hearing Technicians (B4 School Check) and Newborn hearing Screeners (UNHSEIP)[1]. Age of detection profiles from the NZDNDB do show a peak around 4-5 years of age which is assumed to be due to the B4 School screening check[1]. There are also concerns about accessibility of an ad-hoc referral system for deprived families, which may exacerbate social inequalities.

10.7 Are there known harms from screening for hearing impairments in children aged 0-5 years?

Referrals from school entry programmes have minimal to no negative impact on families[34]. However hearing screening programmes can potentially place burden on services, potentially slowing down diagnosis[34]. Note that this is not necessarily the case with hearing screening programmes more likely reducing (false positive) referrals[34].

Additionally screening programmes that have high false positive rates can undermine parental belief in screening accuracy and compliance in diagnostic appointment attendance[18].

As with any screening programme, hearing screening has the potential to increase societal inequalities if not managed carefully: the middle classes tend to make more use of them and interact more positively with the healthcare system while the higher deprivation people are more likely not to engage as effectively, so not gain benefits and thus societal inequalities are exacerbated. Targeting of the screen on at-risk and/or higher deprivation communities, or making sure that coverage is really universal (i.e. 100% uptake) and that there are properly funded and pro-active follow-up procedures for referrals are key approaches to mitigating this risk, but need adequate funding and a properly aligned screening system.

10.8 What interventions or additional support for hearing are effective following early detection?

Following detection there are multiple pathways to support a child with hearing loss. The approach depends on the type of loss, degree of hearing loss, whether the loss is bilateral or unilateral, and the home and educational environment.

The primary cause of hearing loss for the target age group is Otitis Media with Effusion. Management varies depending on whether the effusion is persistent. In most cases OME spontaneously resolves however for some cases active intervention is required to minimise long term detrimental effects. Interventions may include ventilation tube insertion, antibiotics, and ear drum repair; for chronic and longstanding disease, invasive operations may be required (e.g. mastoidectomy).

For sensorineural hearing losses effective management again depends on the degree of hearing loss but options include: speech language therapy, class room sound field systems or personal FM systems, hearing aids, preferential seating and other environmental and behavioural modifications, sign language, and enrolment in a deaf school, cochlear implantation, and auditory verbal therapy.
For Auditory Processing Disorders, there are a range of treatment options including hearing aids and behavioural training and environmental modifications.

10.9 Does early intervention lead to significant improvements later in childhood/ adolescence?

It has been well established that early detection and intervention leads to improved speech and language outcomes for even mild hearing losses[60]. Certainly the benefit of intervention becomes more obvious as the severity of hearing loss increases[61].

There is some debate within the literature regarding the impact of intervention for minimal and more mild losses; as well as unilateral losses. Of note is a cross sectional study which assessed 6581 children in 80 schools in Melbourne[62]. The study found 39 children (0.59%) with a slight sensorineural loss (16 – 25 dB HL) and 16 children (0.24%) with mild sensorineural loss (26 – 40 dB HL) in the better ear; a total of 55 children (0.88% or approximately nine per thousand)[62]. It found no strong evidence that slight/mild bilateral sensorineural hearing loss adversely affect language, reading, behaviour or health-related quality of life[62]. This study is a significant addition to the literature as unlike many other investigations into intervention impact (which have recruited from clinical populations); it has no opportunity for sampling bias. Furthermore given the higher prevalence of mild hearing loss in the Māori population

(see below) research in New Zealand would be of benefit to assess what the rates are here and whether slight to mild losses are associated with negative outcomes.

Conductive losses were excluded from this study. Children with a history of conductive loss (as a result of otitis media with effusion) are more likely to present with spatial processing disorder, a form of auditory processing disorder[63]. Interventions exist for auditory processing disorder however accessing such interventions may be challenging due to a relatively small number of clinicians specialising in this area.

Regarding chronic otitis media, earlier intervention leads to less complications later in life, however care needs to be taken to ensure that screening services do not needlessly refer cases that will likely spontaneously resolve which can overwhelm diagnostic services. A Cochrane review found no clinically significant benefits to language and behaviour outcomes of screening and early treatment of OME in the first four years of life for the general population[64]. The reviewers did take care to note however that the findings may not be the same for high risk populations where incidence of OME complications is higher and early intervention may reduce complication severity which includes the same as that for hearing loss[64,65].

10.10 What do we know from a Māori and Pacific knowledge basis about screening in this domain?

A study on 485 South Auckland children aged 2-3yrs that attended a screening recall due to a problem with their newborn hearing screen found Māori and Pacific ethnicity was significantly associated with hearing loss[66]. They concluded that "there is a high proportion of children in South Auckland with unsuspected hearing loss" and that "a different approach to screening is warranted for this population with high rates or middle ear disease at age 3"[66]. Pacific children have a higher incidence of ear disease even at 2 years of age[67], and this increased incidence of disease may account for more disabling losses and higher fail rates for the B4 school check.

More Māori and Pasifika children fail the B4 School check than any other ethnic groups[66]. Young Māori have a higher incidence of hearing loss than NZ Europeans and their hearing losses are more likely to be mild-moderate and bilateral[1,68]. This is an important finding as generally speaking mild losses are less likely to be detected[66,69] and their impacts on learning are less likely to be understood. This influences how families treat the condition and consequently support interventions such as hearing aids. Therefore it has been recommended that screening programmes must be supported by good community education programmes and appropriate habilitation options for families[68]. This is particularly relevant because Māori and Pacific Island children appear to be under-represented for otitis media hospitalisations and have higher rates of non-attendance at ENT out-patient clinics[70].

Coverage rates for the B4 School check are poorest for Pasifika children with 10.4% not checked compared to 4.8% of New Zealand Europeans and 0.2% of Māori children, (note this is a significant improvement in coverage for Māori from a high of 28% not checked in 2010/11)[1].

10.11 Summary of Findings and Graded Evaluations

- No reliable NZ specific prevalence data or data regarding current efficacy of the B4 school check as it stands could be found. If looking to make changes to current screening programmes improving reporting and obtaining efficacy data would be useful.
- Targeted screening of at-risk populations for OME should be investigated further (at-risk being higher deprivation regions, and Pacific and Māori populations), with implementation most likely done at 3 years of age.
- Regarding level of screening (pass rates) it may be acceptable to exclude minimal and smaller losses for school aged children. However, consideration must be given to at risk populations including Māori and Pacific peoples. Notably Māori who tend to have a higher prevalence of mild sensorineural hearing loss which already tends to be detected or occur later in childhood. Such a shift in approach therefore needs to be considered carefully and research conducted to determine the prevalence and impacts in New Zealand.
- Recommendations around school entry and current B4 school programme are difficult to make without the prevalence and current efficacy data. From international data there is good evidence to shift to DPOAE screening with tympanometry and puretone sweep testing as backup for a DPOAE refer result to reduce the rate of false positive referrals.
- For other populations (developmentally delayed) DPOAE screening is clearly the best option and should be implemented.
- School age screening using Sound Scouts may be an appropriate tool, perhaps at school entry* and at year 3 and 5 (as recommended on the SoundSkills website) this could be tied into the academic health studies in later years, and likely has minimal cost and can be implemented easily. However caution is recommended at this stage as all data currently available is from a single study. Furthermore, the test is currently self-administered, and a protocol and support system would have to be set up to ensure equitability of outcomes across the community.

Table 10.4. Graded evaluation of screening tools and associated recommendations for policy and practice.

Screening Tool	Grade	Estimated net benefit	Level of certainty	Recommendation
Manual Pure-Tone Screen	B	Substantial	Moderate	This tool is widely internationally. There are concerns however regarding its reliability in younger populations and in background noise.
Automatic (Phone App) Pure-Tone Screen	C	Moderate	Moderate	This approach may be useful for school children age 6+; the main advantage is that as opposed to manual testing screeners require less training.
Digits in Noise Test	I	Insufficient evidence	Low	May be applicable for screening older children 9 years and above, currently not enough evidence to recommend for younger children. Primary benefit is that this type of test can be performed as an online test. Possible limitation is that it has poor sensitivity for conductive losses.
TEOAEs	C	Moderate	Moderate	TEOAES are quick and sensitive to moderate hearing losses but are not sensitive to minimal hearing losses and perform poorly in noisy environments.
DPOAEs	B	Substantial	Moderate	DPOAEs are widely used for screening hearing in children 3 years or younger. They are fast and require minimal patient cooperation. It is a sensitive screening tool, however false positive rates may be higher than pure-tone screening. DPOAE testing may be a good first line screen for all ages, with a second screen of manual pure-tone and tympanometry for those who get a refer result.
Game Based Screening (Sound Scouts)	B	Substantial	Moderate-Low	This tool has been made available online in Australia, and is suitable for ages 4.5 and above in developmentally normal children. Sensitivity and specificity is equivalent to published data for both DPOAEs and the Pure-Tone Screen if the goal is to detect slight and mild losses, and even better for larger losses. It requires a longer time to conduct the testing, and does not requires an adult to supervise but not specialist training to administer. All data for this approach comes from a single study.
Questionnaires	D	Nil	High	Not useful for this population
Auditory Evoked Potentials	D	Nil	High	Not useful for this population

Grade: A, B, C, D, or I.
Estimated net benefit: substantial, moderate, small, nil or harmful, or insufficient (evidence).
Level of certainty: high, moderate, or low.
For more detailed explanation see Supplementary Information - Grade definitions and levels of certainty.

Table 10.5. Graded evaluation of interventions and associated recommendations for policy and practice.

Intervention	Grade	Estimated net benefit	Level of certainty	Recommendation
Treatments for persistent middle ear disease	A	Substantial	High	This intervention should be provided for every child who needs it, dependent upon history and extent of disease, this may involve surgery and be dependent upon surgical waiting lists.
Hearing Aids / Cochlear Implants	A	Substantial	High	This intervention should be provided for every child who needs it. Specific intervention depends upon several factors (degree of loss, speech recognition performance, performance in school), and is decided upon by professionals and parents
FM Systems / Soundfield Systems	A	Substantial	High	This intervention should be provided for every child who needs it. Application is dependent upon a child's hearing performance in the classroom environment, and also the teacher's and student's willingness to use the devices.
Sign language / Deaf School	A	Substantial	High	This intervention should be provided for every child who needs it. For some children tradition amplification of cochlear implants are not an option, or parents may choose this mode of communication.
Auditory Verbal Therapy / Speech language therapy	A	Substantial	High	This intervention should be provided for every child who needs it. In cases where children have a significant hearing loss, or a late diagnosis, speech language therapy is usually required to help them make the most of language and the habilitation devices they are using.
Behavioural and Environmental Modifications	A	Moderate	High	This intervention should be provided for every child who needs it. Simple environmental and behavioural modifications (e.g. acoustic tiling, sitting closer to the target talker) help all children with hearing loss. They are low cost and generally easy to implement.

Grade: A, B, C, D, or I.
Estimated net benefit: substantial, moderate, small, nil or harmful, or insufficient (evidence).
Level of certainty: high, moderate, or low.
For more detailed explanation see Supplementary Information - Grade definitions and levels of certainty.

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Supplementary Information - Grade definitions and levels of certainty

Table S1. Grade definitions for screening tools and interventions

Adapted with permission from the U.S. Preventive Services Task Force 2012.[i]

Grade	Definition	Recommendation for policy and practice
A	• The authors recommend this screening tool/intervention. • There is high certainty that the net benefit is substantial.	• This screening tool/intervention should be offered or provided.
B	• The authors recommend the screening tool/intervention. • There is high certainty that the net benefit is moderate, or there is moderate certainty that the net benefit is moderate to substantial.	• This screening tool/intervention should be offered or provided.
C	• The authors recommend selectively offering or providing this screening tool/intervention to patients based on professional judgment and patient preferences. • There is at least moderate certainty that the net benefit is small.	• This screening tool/intervention should be provided for selected patients depending on individual circumstances.
D	• The authors recommend against this screening tool/intervention. • There is moderate or high certainty that the screening tool/ intervention has no net benefit or that the harms outweigh the benefits.	• The authors discourage the use of this screening tool/intervention.
I	• The authors conclude that the current evidence is insufficient to assess the balance of benefits and harms of the screening tool/intervention. • Evidence is lacking, of poor quality, or conflicting, and the balance of benefits and harms cannot be determined.	• If the screening tool/intervention is offered, patients should understand the uncertainty about the balance of benefits and harms.

Table S2. Levels of certainty regarding net benefit

Adapted with permission from the U.S. Preventive Services Task Force 2012 [1].

Level Of Certainty	Description
High	• The available evidence usually includes consistent results from well-designed, well-conducted studies in representative populations. • These studies assess the effects of the preventive service on health outcomes. • This conclusion is therefore unlikely to be strongly affected by the results of future studies.
Moderate	• The available evidence is sufficient to determine the effects of the preventive service on health outcomes, but confidence in the estimate is constrained by such factors as: – the number, size, or quality of individual studies; – inconsistency of findings across studies; – limited generalizability of findings to routine practice; – lack of coherence in the chain of evidence. • As more information becomes available, the magnitude or direction of the observed effect could change, and this change may be large enough to alter the conclusion(s).
Low	• The available evidence is insufficient to assess effects on health outcomes, because of: – the limited number and/or size of studies; – important flaws in study design and/or methods; – inconsistency of findings across individual studies; – gaps in the chain of evidence; – findings not generalizable to routine practice; – lack of information on important health outcomes. • More information may allow estimation of effects on health outcomes.

[i] https://www.uspreventiveservicestaskforce.org/Page/Name/grade-definitions